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Original article

Treatment of dystonia in extensor hallucis longus and digitorum muscles with neurotomy of the branches of the deep fibular nerve: Preliminary results

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ABSTRACT

Introduction: Dystonia in extensor hallucis and/or digitorum muscles can be observed in pyramidal and extrapyramidal lesions and results in pain in these toes, spontaneous or when walking, problems and discomfort when putting on shoes and socks, and cutaneous lesions on the toes. The objective of this study was to assess the efficacy and safety of deep fibular nerve neurotomy for the extensor hallucis longus (EHL) and/or the extensor digitorum longus (EDL) branches in the treatment of extension dystonia of the hallux and/or other toes.
Patients and methods: A deep fibular nerve neurotomy was performed in 20 patients ($n = 19$ for the EHL, $n = 6$ for the EDL). We retrospectively analyzed the treatment's efficacy and safety and assessed the patients' self-reported improvement and overall treatment satisfaction.
Results: Dystonia totally disappeared in 15 cases (75%); it persisted at a minimal level in the other patients. The patients reported a decrease in pain ($P < 0.01$) and fewer difficulties putting on shoes and socks ($P < 0.001$) and had a high median level of satisfaction (8.5/10). Adverse effects were rare and transient. The identification of the nerve branches was sometimes difficult.
Discussion: Deep fibular nerve neurotomy for the EHL and/or EDL branches seems to be an effective treatment for extension dystonia of the hallux and/or other toes and its consequences for the adult neurological patient. However, these encouraging preliminary results should be confirmed by prospective, longer-term studies.
Level of evidence: IV.

1. Introduction

Extension dystonia of the hallux and/or toes is a classical deformity [1], encountered in two distinct contexts: extrapyramidal deformity [2] and pyramidal lesions secondary to neonatal or acquired brain injury [1,3]. It is related to hypertonia of the extensor hallucis longus (EHL) and/or the extensor digitorum longus (EDL) muscles. These extension spasms can lead to troublesome symptoms such as spontaneous or induced pain (either on the dorsal side of the toe(s) or of their tendon, which, when under tension, can become painful), problems putting on shoes and socks, discomfort when wearing shoes, cutaneous lesions on the dorsal side of the hallux [3–7], as well as problems when walking related to the pain induced and to the instability that can occur at the end of the stance phase [3,5,6].

The hyperextension mechanism always involves the EHL and/or the EDL, which can be hypertonic, retracted, or both. In its isolated form, this deformity involves hyperextension of the metatarsophalangeal (MTP) and the interphalangeals (IPs). If it is associated with tension on the flexor system, it often presents as hyperextension of the MTP and flexion of the IPs. Hyperactivation of the EHL and/or the EDL first of all can be caused by true dystonia of these muscles. When it occurs in the swing phase, it can also be related to a tenodesis effect on the EHL and/or the EDL when the foot drops because of a deficit in the levator muscles. Finally, it can correspond to an attempt to compensate for weakness in the anterior

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tibialis anterior muscle by the extensor digitorum muscles, which are accessory dorsal flexors. In these three cases, the severity of the hyperextension can be aggravated by retraction of the muscles involved.

Over the past few years, botulinum toxin treatment (BT) has proved to be effective in treating dystonia of the EHL in extrapyramidal lesions such as after an acquired brain injury [3–5]. However, this treatment often requires high doses of BT [5], limiting its use in other hypertonic muscle groups, and necessitates repeated injections. Neuro-orthopaedic treatment aims to prevent these two pitfalls. Lengthening the EHL tendon at the muscle–tendon junction is a therapeutic possibility [8,9], all the more indicated in isolated hyperactivity of the extensor muscles when muscle retraction is present, even though to our knowledge the literature reports no specific data on neurological hyperextension of the toes concerning this technique, nor any description of EDL lengthening procedures. In our experience, toe tendon lengthening procedures tend to give inconsistent and transient results, and the degree of lengthening is often difficult to predict (and is therefore a source of under- or over-correction). As for other locations, neurotomies have been shown to be effective in treating muscular hypertonia associated with pyramidal syndrome [10]. We have developed a selective neurotomy technique of the motor branches of the deep fibular nerve (formerly referred to as the anterior tibial nerve) leading to the EHL and the EDL muscles.

In the present study, we analyzed the efficacy and safety of deep fibular nerve neurotomy in adult patients suffering from disabling extension dystonia of the hallus and/or the other toes.

2. Patients and methods

2.1. Patients

In this preliminary open multicenter study, we included patients presenting with dystonia in of the hallux and/or other toes, causing problems (pain, cutaneous lesions on the toes, problems putting on shoes and socks, discomfort when walking), for which surgical treatment had been recommended during a multidisciplinary consultation. This consultation, which grouped physicians in physical and rehabilitation medicine, orthopaedic surgeons, and a neurosurgeon, allowed us to analyze the impact of dystonia of the toes, to select the targets that should profit from neurotomy (EHL and/or the EDL), and obtain the patient's informed consent. We included patients whatever the source of the dystonia, excluding patients suffering from severe language or psychiatric disorders because they would have been unable to take part in a subsequent telephone interview. The study was conducted in compliance with the principles of the Declaration of Helsinki and the patients were requested to provide oral consent to participate in the study.

Deep fibular nerve neurotomy was performed in 20 patients between July 2011 and January 2013 (Table 1). Twelve (60%) were female, and the median age was 52 years (range, 27–64 years). They suffered from stroke (14 patients, 70%), traumatic brain injury (two patients, 10%), cerebral palsy (one patient), craniostenosis (one patient), primary generalized dystonia (one patient), and cerebrotendinous xanthomatosis (one patient). For acquired lesions, the median duration was 9.5 years (range, 2.2–23.1 years). Six patients (30%) had been treated with BT for dystonia of the EHL, with at least partial efficacy for five of them.

2.2. Treatment

Neurotomy for fibular nerve branches that innervate the EHL and/or the EDL was performed under general anesthesia, with the patient in the supine position. A vertical cutaneous incision on the anterolateral side of the leg provided access to the space separating the tibialis anterior medially and the EHL and EDL laterally, making it possible to first approach the deep fibular nerve trunk and to repair the motor branches for both muscles. There was always a common branch for the EHL and EDL, and a varying number of additional branches for each of the muscles (from one to three) [11]. Since the course of the nerve varies between patients, selective fascicular electrical neurostimulation was used to identify motor branches and avoid sensory branches. Once they had been identified, the muscular branches were resected from three-fifths to four-fifths over a distance of 1 cm [12], such that the muscle response to upstream electrical stimulation was clearly reduced.

The neurotomy targeted the EHL branches for 14 patients (70%), the EDL alone for one patient (5%), and both for five patients (25%) (Table 1). It was bilateral in two patients (for the EHL in one case and the EDL in the other). The median follow-up period was 9.5 months (range, 3–21 months). In most cases, the patients underwent other therapeutic procedures in the same surgical time: one tibial nerve neurotomy for the gastrocnemius, the soleus, and tibialis posterior was done in three patients (15%), tendon lengthening in 16 patients (80%), which in 11 (55%) involved the Achilles tendon, 11 the flexor hallucis longus, and 12 the flexor digitorum longus (tenotomy). Two patients (10%) underwent split tibialis anterior transfer, and three (15%) a subtalar arthrodesis.

2.3. Evaluations

A retrospective study was first conducted by comparing the pre- and postoperative consultation data (carried out between 3 and 6 months after the procedure). The location of the dystonia was recorded (hallux, other toes, or both), its severity (0 = absent, 1 = minimal, 2 = moderate, 3 = severe, based on its amplitude and the frequency with which it occurred), its triggering features (permanent or initiated when standing or walking), the problem(s) occurring in both passive functions (spontaneous pain in the dorsal side of the toes or the extensor tendons, problems and discomfort when putting on shoes and socks, toe calluses and ulcers) and when walking (pain). In addition, intra- and postoperative adverse events were systematically recorded.

In the second part of the study, all patients were contacted by telephone in May 2013 by two investigators who were not members of the surgical team (CR and EA) to assess the self-reported changes induced by the neurotomy. The patients were first requested to evaluate their main dystonia-induced complaint before surgery (among those listed above) and to score the overall impairment related to the dystonia when standing and walking before and after the procedure on a numeric scale (from 0 = no impairment to 10 = major impairment). In addition, they were asked to assess the changes over time in dystonia-induced impairment on the Global Assessment Scale (GAS) [13], which uses a Likert scale from −4 (major deterioration) through 0 (no change) to 4 (major improvement). Finally, the patients were requested to estimate their overall satisfaction with the treatment of the EHL and/or the EDL dystonia using a numeric scale (from 0 = not satisfied at all to 10 = very satisfied).

2.4. Statistical analysis

Continuous variables were expressed as median and range, the ordinal variables as median and interquartile interval. The preoperative and postoperative comparisons were made using a Chi^2 test for the qualitative variables and a Wilcoxon test for the quantitative variables. The significance threshold was set at 0.05 (bilateral significance). The statistical analyses were performed using SPSS software (version 20, SPSS Inc., Chicago, IL, USA).

Table 1
Description of the population.

Patient	Age (years)	Gender	Diagnosis	Time since lesion (years)	Treatment	Side	Follow-up period (months)
1	52	F	Stroke	23.1	EHL	Left	20
2	64	M	Stroke	5.6	EHL	Left	14
3	54	F	Stroke	9.6	EHL	Left	14
4	49	M	Stroke	1.9	EHL	Left	14
5	35	F	Stroke	21.6	EHL	Left	14
6	63	F	Stroke	12.8	EHL + EDL	Left	11
7	61	F	Stroke	8.2	EHL + EDL	Left	10
8	45	M	Stroke	2.9	EDL	Right	10
9	60	M	Stroke	2.2	EHL	Left	9
10	52	M	Stroke	5.1	EHL	Left	8
11	49	F	Stroke	2.9	EHL	Left	8
12	57	F	Stroke	8.8	EHL	Left	5
13	59	F	Stroke	13.4	EHL	Left	4
14	58	M	Stroke	3.6	EHL	Right	3
15	54	M	TBI	10.0	EHL + EDL	Left	11
16	46	F	TBI	21.2	EHL + EDL	Left	3
17	27	M	Cerebral palsy	–	EHL	Right	7
18	39	F	Cerebrotendinous xanthomatosis	–	EHL + EDL	Bilateral	3
19	52	F	Craniostenosis	–	EHL	Left	12
20	37	F	Generalized dystonia	–	EHL	Bilateral	8

F: female; M: male; TBI: traumatic brain injury; EHL: extensor hallucis longus; EDL: extensor digitorum longus.

Table 2
Pre-/postsurgery changes in the severity of dystonia and its triggering features (based on clinical examination, retrospective).

	Preop	Postop
Triggering features		
Permanent	2 (10.5%)	0
Triggered	18 (90%)	5 (25%)
When standing	12 (60%)	2 (10%)
When walking	18 (90%)	3 (15%)
Dystonia severity		
Mild	3 (15%)	5 (25%)
Moderate	13 (65%)	0
Severe	4 (20%)	0

Data are the number of patients (%). Postop: 3–6 months after procedure.

3. Results

3.1. Symptoms and data from the clinical examination

Data showing the change over time in the dystonia's severity and triggering features are summarized in Table 2. Before the surgery, dystonia most often triggered when standing or walking, it was permanent in two cases of dystonia of the EHL. The clinical severity of the dystonia was moderate to severe in most cases. None of the patients presented contracture of the EHL and/or the EDL. After treatment, complete disappearance of the dystonia was observed in 15 cases (75%), it persisted in five patients but always with mild severity. No flexion claw toes were observed after the surgery.

Deep fibular nerve neurotomy was associated with a marked reduction in the disabling consequences of dystonia identified prior to treatment (Table 3). This was particularly true for the most frequent complaints (spontaneous pain, difficulties and discomfort putting on shoes and socks). All the patients could walk before the procedure, which led to no notable change in walking conditions (assistive devices, orthotics needs, shoes).

Table 3
Pre-/postsurgery changes in the disabling consequences of dystonia (based on data in medical records, retrospective).

Discomfort	Preop	Postop
Spontaneous pain	8 (40%)	1 (5%)
Pain when walking	16 (80%)	1 (5%)
Difficulties/discomfort wearing socks/shoes	18 (90%)	3 (15%)
Cutaneous lesions	5 (25%)	1 (5%)

Data are the number of patients (%). Postop: 3–6 months after procedure.

3.2. Subjective assessment

Before treatment, the patients' main complaint related to their dystonia was the difficulties and discomfort when putting on shoes and socks ($n = 11$, 55%), pain in the toes involved in the dystonia when standing or walking ($n = 4$, 20%), poor balance when walking ($n = 4$, 20%), and spontaneous pain in the toes ($n = 1$).

After the intervention, the overall impairment related to standing and walking was substantially improved ($P < 0.01$ and 0.001, respectively) (Table 4). Likewise, all the patients' complaints were relieved to a fair degree after surgery, notably for discomfort and difficulties when putting on and wearing shoes and socks and pain when walking. Only the improvement in cutaneous lesions on the toes, although quantitatively high, remained nonsignificant. The patients' overall satisfaction with the treatment of dystonia of the EHL and/or the EDL reached a median of 8.5 (range, 5–10).

3.3. Peri- and postoperative adverse events

Four patients (20%) presented a transient peri- or postoperative adverse event. One patient presented a delay in healing (< 2 months) and three patients (15%) reported transient neurogenic pain (lasting no more than 6 weeks) or dysesthesia of the dorsum of the foot or around the surgical scar. In addition, the procedure was

Table 4
Changes in subjective evaluations of the different problems presented by the patients, as related to the dystonia (data from telephone interview).

	Preop	Postop	*P*
Overall impairment (0–10)			
Standing	4 (8)	0	< 0.01
Walking	8 (3.25)	0.5 (1.25)	< 0.001
Subjective changes (GAS −4; 4)			
Spontaneous pain ($n = 8$)		3 (1)	< 0.01
Pain when walking ($n = 16$)		3 (1)	< 0.001
Difficulties/discomfort wearing socks/shoes ($n = 18$)		3 (1)	< 0.001
Cutaneous lesions ($n = 5$)		3 (3)	0.066

Data are median and interquartile interval values. GAS: Global Assessment Scale; Postop: at the telephone interview (median, 9.5 months after the procedure [range, 3–21 months]).

not totally completed in three patients: all the branches could not be repaired in two and in one the neurostimulator used was defective. In this patient, the BT treatment had to be resumed. We noted no deterioration of motor command of the dorsal flexor muscles in the postoperative period.

4. Discussion

This study is the first to evaluate the efficacy and safety of deep fibular nerve neurotomy for the EHL and EDL in the treatment of extension dystonia of the hallux and the toes. The results of this open retrospective study seem to show substantial efficacy in both the clinical characteristics of dystonia (triggering features, severity) and its impact. Patient satisfaction was high. Adverse events were rare and transient, although the perioperative identification of nerve branches was sometimes difficult.

The data from the current study on the impact of dystonia of the EHL and/or the EDL confirm the data reported in the literature [3–5]. This was observed mainly on passive functions, i.e., in problems putting on and wearing socks and shoes, spontaneous or walking-induced pain in the toes, and the onset of cutaneous lesions of the toes. No data exist in the literature on the prevalence and effects of EDL dystonia. In our population, it was less frequent than dystonia of the EHL and, in all but one case, the two were associated.

We report a nearly 75% rate of complete disappearance of the dystonia after neurotomy, whereas residual dystonia was always mild. BT treatment improved the condition similarly, without necessarily complete disappearance [4]. The effects of dystonia on putting on shoes and socks, pain, and walking were substantially reduced, whether assessed by the examiner or the patient. There was also improvement in cutaneous lesions, but it remained nonsignificant; this could be explained by the small number of patients concerned and the fact that one patient had considerable co-morbidities and retained cutaneous lesions despite the substantial efficacy on the dystonia. We consider that the patient's self-assessment is an important part of the evaluation protocol. The level of patient satisfaction was high, even when an adverse effect occurred.

In our experience, deep fibular nerve neurotomy for branches leading to the EHL and/or the EDL is most often combined with a concomitant surgical procedure, or sometimes performed after the treatment of a more global deformity of the distal extremity of the lower limb, most often an equinovarus deformity. Only one patient presenting with generalized dystonia was treated with deep fibular neurotomy alone. As has been demonstrated for equinovarus deformity, neuro-orthopaedic treatment (in particular neurotomy) gives long-lasting, more effective results than BTI does [14–16]. It also allows one to save BT for other hypertonic muscles. This is particularly pertinent for EHL dystonia, which is usually more severe than dystonia of the upper limb [17] and requires high doses of BT [5].

Adverse effects occurred in 20% of the patients, but they were always transient and benign and did not impact the level of patient satisfaction at the intermediate term. The fact that the procedure could not be completed in three patients reflects the difficulties possibly encountered in identifying the different motor branches of the EHL and/or the EDL. Even though a common trunk for the EHL and the EDL is always present, up to three additional branches may be present for each muscle, which could sometimes be difficult to identify. Precise, extensive dissection of the deep fibular nerve is therefore required, which leaves quite a long scar, and the patient must be advised of this.

This study suffers from a number of limitations. In addition to its retrospective design and the absence of a control group, the study population was heterogeneous and included a high proportion of patients with traumatic brain injuries compared to patients presenting injury to the basal ganglia. Yet the physiopathology of dystonia of the toes is not strictly identical in the two cases [18], which limits the extrapolation of the results. The subjectivity of the evaluation criterion of frequency and severity of dystonic movements can also be regretted, but today no specific tool adapted to dystonia of the toes is available. Finally, given the relatively short follow-up period, the long-term effects of deep fibular nerve neurotomy remains to be established, particularly given that the physiopathology of toe dystonia is not identical to that commonly seen in spasticity.

5. Conclusion

Deep fibular nerve neurotomy of the branches leading to the EHL and/or EDL could be of interest for relieving hyperextension of the hallux and/or other toes and reducing the latter's disabling consequences. Adverse events were rare and transient, but identification of all the motor branches was sometimes difficult. These encouraging preliminary results must now be confirmed in a large long-term prospective study.

Disclosure of interest

The authors declare that they have no conflicts of interest concerning this article.

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